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Case report

Spontaneous intra-parotid pseudoaneurysm of the external carotid artery

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ABSTRACT

Introduction: The authors report a case of spontaneous intra-parotid pseudoaneurysm of the external carotid artery, never previously described in the literature.
Case report: A 59-year-old woman presented with a palpable, non-pulsatile, 2 cm retromandibular left parotid mass with no triggering factors. This mass was considered to be a mixed tumour, although it did not present the typical features on MRI. At surgery, the lesion in the lower pole of the parotid gland was found to be pulsatile, in favour of pseudoaneurysm of the external carotid artery. The external carotid artery was ligated to remove the lesion. Definitive histological examination confirmed the diagnosis of pseudoaneurysm.
Discussion: Pseudoaneurysms of extracranial arteries are rare, most commonly involve the internal carotid artery and are essentially secondary to trauma. Review of MRI images demonstrated hyperintense arterial blood flow within the hypointense mass on T2-weighted sequences. The presence of this sign must therefore be investigated in the context of atypical images of a parotid mass. Although rare, and despite the absence of trauma, the diagnosis of pseudoaneurysm of the external carotid artery or one of its branches should therefore be considered in the presence of an atypical parotid mass.
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1. Introduction

Clinicians must be familiar with the various causes of parotid mass and must therefore be aware of the rare causes of enlarged parotid gland that can be a source of diagnostic errors. Appropriate complementary investigations must be performed to ensure adapted therapeutic management.

Pseudoaneurysms of the external carotid artery or its branches are rare [1] and are often secondary to trauma. The spontaneous nature of the intra-parotid pseudoaneurysm of the external carotid artery reported here makes this case a unique case that has not been previously described in the literature.

2. Case report

We report the case of a 59-year-old woman who presented with a left parotid mass in March 2014. She had been previously operated in 2003 for a cystic lesion arising from the salivary ducts of the right parotid gland, with no signs of malignancy.

Initial clinical examination demonstrated a palpable, deep retromandibular, non-pulsatile mass, measuring 2 cm in diameter. MRI demonstrated a heterogeneous nodular formation, 20 mm in diameter, in the deep lobe of the left parotid gland with regular contours and no infiltration of the adjacent parenchyma. This T1-hyperintense and T2-hypointense lesion showed gadolinium enhancement on the T1-weighted sequence (Fig. 1). Although not presenting the typical signs, the radiologists initially concluded on a diagnosis of mixed tumour.

Surgical resection consisted of subcutaneous detachment and dissection of the superficial musculoaponeurotic system (SMAS) with dissection of the parotid gland, comprising identification and sparing of the facial nerve. The deep lesion was situated in the lower pole of the parotid, below the cervicofacial branch of the facial nerve, 1 cm below the bifurcation of the two main branches of the facial nerve. The mass did not present any signs of malignancy but was intensely pulsatile during the operation. Following dissection of adjacent structures, the lesion appeared to be an intra-parotid pseudoaneurysm of the external carotid artery arising from the anterior wall of the artery. It was decided to remove the lesion, requiring ligation of the external carotid artery proximal and distal to the lesion to allow resection. Intraoperative frozen section examination confirmed the vascular nature of the lesion with no criteria

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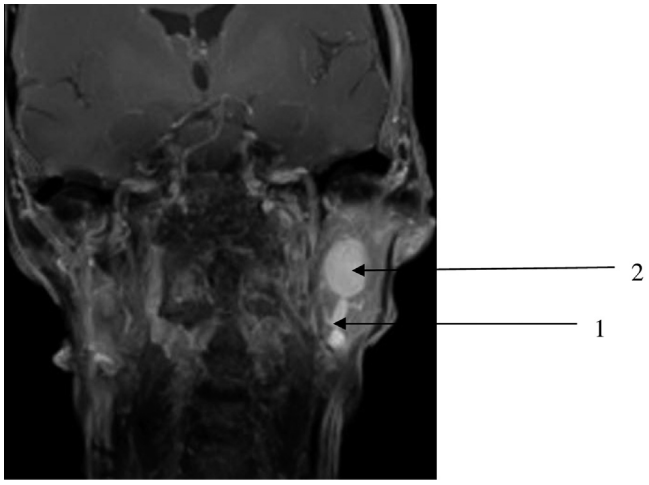

Fig. 1. MRI T1 coronal section: visualisation of the external carotid artery (1) and the pseudoaneurysm mimicking an intra-parotid mass (2).

of malignancy. Facial nerve-sparing parotidectomy was therefore not continued.

Definitive histological examination confirmed the diagnosis of pseudoaneurysm of the external carotid artery.

3. Discussion

Pseudoaneurysms of extracranial arteries are rare and usually post-traumatic, commonly involving the internal carotid artery [2]. Only 13 cases involving the external carotid artery or its branches have been reported in the literature [2,3]. The most common forms of pseudoaneurysm involve the superficial temporal artery and are essentially post-traumatic or iatrogenic [4–6]. Spontaneous pseudoaneurysm is an extremely rare diagnosis.

An intra-parotid aneurysm can present as a parotid mass. Only 7 cases have been reported in the literature [3]: 4 cases involving the intra-parotid segment of the external carotid artery, 2 cases involving the superficial temporal artery and 1 case involving the posterior auricular artery [7]. All of these cases were secondary to an identified triggering factor.

The case reported here presented clinically as a parotid mass. The usually pulsatile nature of aneurysms and pseudoaneurysms was absent on initial clinical examination of our patient. It should be noted that mixed tumours do not classically present with the radiological signs observed in this case. However, despite the absence of typical features, and in a clinical context of pain, further complementary investigations were not performed and it was decided to perform surgical exploration of this lesion. The diagnosis of pseudoaneurysm of the external carotid artery was established intraoperatively based on the anatomical features of the lesion. Postoperative review of MR images revealed T2-hyperintense intra-aneurysmal arterial flow within a T2-hypointense mass (Figs. 2 and 3). This sign, not previously reported in the literature, must therefore be investigated in any case of T1-hyperintense, T2-hypointense mass.

Cases of spontaneous pseudoaneurysms induced by repeated Valsalva manoeuvres have been reported in the literature [8], but this mechanism was not observed in our case. This case highlights the importance of keeping in mind rare causes of intra-parotid lesions, but especially the importance of complementary investigations (ultrasound, CT, MRI) and correct interpretation of these examinations before performing fine-needle aspiration or surgical biopsy. Imaging modalities such as high-resolution MRI or contrast-enhanced multi-array CT allow

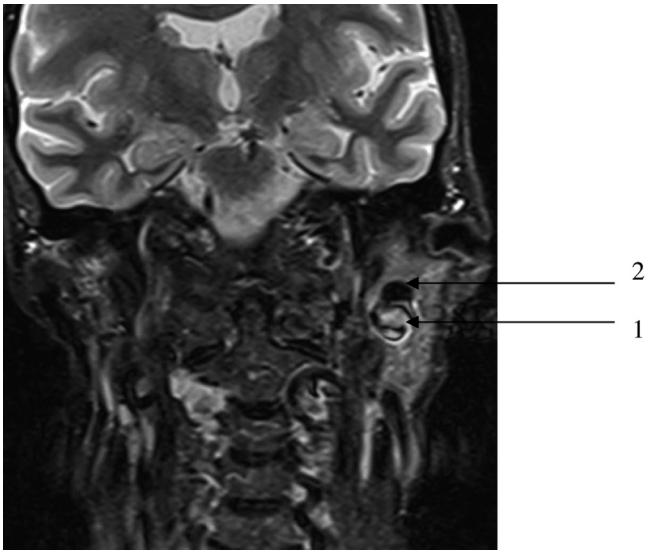

Fig. 2. MRI T2 coronal section: visualisation of T2-hyperintense arterial blood flow (1) within a T2-hypointense parotid mass (2).

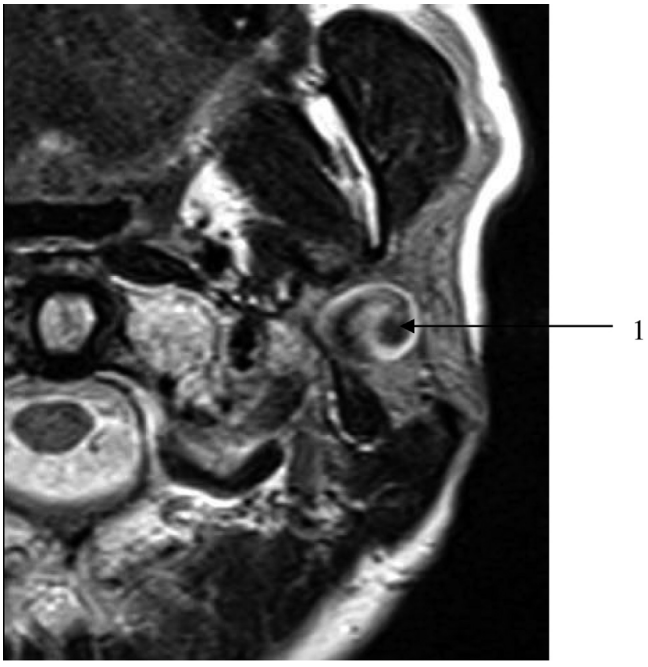

Fig. 3. MRI T2 transverse section: visualisation of hyperintense arterial blood flow within a T2-hypointense mass (1).

precise visualisation of anatomical features and also allow various angiographic sequences [9]. Dynamic MRI sequences and diffusion-weighted with measurement of the ADC also allow improved performance of MRI to distinguish benign and malignant parotid tumours and to characterize the histological type of benign tumours [10]. In the case reported here, the use of these techniques may have allowed a preoperative diagnosis of pseudoaneurysm. However Doppler ultrasound can also be used to visualise the various blood flows (luminal and aneurysmal).

The place of fine-needle aspiration cytology remains controversial. Fakhry et al. [11] showed that fine-needle aspiration cytology is a reliable examination that is able to provide valuable preoperative

diagnostic information with a sensitivity of 80% and a specificity of 89.5% in the context of malignant parotid tumours. Fine-needle aspiration cytology of intra-parotid pseudoaneurysm would not have been possible and would certainly have led to an increase in the size of the lesion. Complementary investigations are currently underway to identify the cause of this pseudoaneurysm.

The management of extracranial pseudoaneurysm depends on various factors: compression of adjacent structures, increasing dimensions of the lesion, associated symptoms (proximity of the facial nerve) and the patient's preference. The various treatment options comprise watchful waiting, surgical resection, sclerotherapy and embolization [3,5].

In the present case, treatment consisted of surgical resection with ligation of the external carotid artery. However, this procedure was performed without a preoperative plan, as the diagnosis of aneurysm or pseudoaneurysm had not been considered. It therefore appears essential to consider the possibility of this disease in the presence of an atypical lesion on MRI before performing any invasive procedure.

4. Conclusion

Although rare, and despite the absence of trauma, it is important to consider the possibility of pseudoaneurysm of the external carotid artery or one of its branches in the presence of atypical imaging signs of an intra-parotid tumour, thereby allowing a more complete complementary assessment in order to propose appropriate management.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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